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Developmental trajectories of pitch-related music skills in children with Williams
syndrome

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Abstract

The study of music cognition in Williams syndrome (WS) has resulted in theoretical debates regarding cognitive modularity and development. However, no research has previously investigated the development of music skills in this population. In this study, we used the cross-sectional developmental trajectories approach to assess the development of pitch-related music skills in children with WS compared with typically developing (TD) peers. Thus, we evaluated the role of change over time on pitch-related music skills and the developmental relationships between music skills and different cognitive areas. In the TD children, the pitch-related music skills improved with chronological age and cognitive development. In the children with WS, developmental relationships were only found between several pitch-related music skills and specific cognitive processes. We also found non-systematic relationships between chronological age and the pitch-related music skills, stabilization in the level reached in music when cognitive development was considered, and uneven associations between cognitive and music skills. In addition, the TD and WS groups differed in their patterns of pitch-related music skill development. These results suggest that the development of pitch-related music skills in children with WS is atypical. Our findings stand in contrast with the views that claim innate modularity for music in WS; rather, they are consistent with neuroconstructivist accounts.

Key words: Williams syndrome; pitch-related music skills; developmental trajectories; atypical development

What this paper adds?

To our knowledge, this is the first research on the development of music skills in Williams syndrome (WS). The cross-sectional developmental trajectories approach was used to study pitch-related music skills in children with WS. We found a set of atypicalities in the development of such skills. Our results contribute to the theoretical debates regarding music cognition in WS. The findings are consistent with neuroconstructivist accounts and represent further evidence against the nativist views of modularity for music in WS.

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1. Introduction

*1.1. The theoretical relevance of studying Williams syndrome: The case of music
cognition*

Williams syndrome (WS) is a genetic neurodevelopmental disorder caused by a microdeletion on 7q.1123 (Ewart et al., 1993); WS occurs in only 1:7500 live births (Strømme, Bjørnstad, & Ramstad, 2002). Individuals with WS exhibit intellectual disability and a cognitive profile traditionally described as presenting both relatively good skills in music, language, face processing, and phonological memory and severe difficulties in visuospatial construction, numerical cognition, planning, and problem solving (e.g., Bellugi, Lichtenberger, Jones, Lai, & George, 2000; Levitin et al., 2004; Mervis et al., 2000). Studies of this uneven cognitive profile have contributed to debates regarding cognitive modularity and development. Thus, WS has been used to support both nativist and neuroconstructivist theoretical views in psychology (Karmiloff-Smith, 2009). While the nativist approach claims the existence of innate and independently functioning modules (Pinker, 1991), the neuroconstructivist view argues against innate pre-specification and emphasizes the role of development and the interaction of multiple levels of explanation when considering phenotypical outcomes (Karmiloff-Smith, 1998a; Mareschal et al., 2007).

This divergence can be illustrated in the field of music cognition. The first reports on the topic described individuals with WS as having preserved and exceptional music skills (see Lenhoff, Perales, & Hickok, 2001a, 2001b, and Levitin & Bellugi, 1998, regarding absolute pitch and rhythm reproduction skills, respectively). From nativist accounts, the fact that individuals with WS seemed to show high music skills

despite their intellectual disability was interpreted as evidence that music represents an innate module independent of general cognition (Levitin & Bellugi, 1998; Lenhoff et al., 2001a, 2001b). Along the same lines but more recently, research on auditory processing in WS has led to claim that the syndrome offers evidence in favor of the idea that musical talent represents an innate predisposition (Wengenroth, Blatow, Bendszus, & Schneider, 2010). However, in contrast with this view but in agreement with neuroconstructivist accounts, other research has argued that the music skills of individuals with WS are affected by their cognitive deficits and are therefore impaired (Martínez-Castilla, Sotillo, & Campos, 2011). This would involve that music is not informationally encapsulated, which in turn suggests that music cannot be considered a module in WS. It has also been claimed that music shares processing mechanisms with other cognitive domains in this population (Martínez-Castilla & Sotillo, 2014). Furthermore, it has been suggested that even when individuals with WS reach proficient music skills, these skills may follow developmental pathways different from those observed in typically developing (TD) individuals (Elsabbagh, Cohen, & Karmiloff-Smith, 2010).

1.2. The role of pitch in music

In the investigation of music cognition, it is important to consider that pitch structure is a highly relevant factor in musical organization and is crucial to music appreciation (Krumhansl & Shepard, 1979; Trehub, Cohen, Thorpe, & Morrongiello, 1986). Pitch, or tone, is the psychological correlate of the acoustic parameter of frequency (Calvo-Manzano, 1991). Individual pitches may form melodies when sequentially presented or chords when simultaneously presented (Cuddy & Badertscher, 1987; Krumhansl, 2004). Regardless of sequential or simultaneous presentations, the ratios between the pitch frequencies form consonances (simple ratios; e.g., 2:1, the

octave interval) and dissonances (complex ratios; e.g., 16:15, the minor second interval) (Hannon & Trainor, 2007; Krumhansl, 2004). In turn, consonance and dissonance discrimination provides a bootstrap into the pitch structure of a musical system, including the Western pitch organization or tonality (Hannon & Trainor, 2007; Trainor, Tsang, & Cheung, 2002). In a broad sense, tonality “involves a hierarchically organized system of pitch relationships” (Cuddy & Badertscher, 1987; p. 609). More specifically, tonality is synonymous with key and defines the musical scale via the establishment of a referential pitch, i.e., the tonic (e.g., C in the key of C major) (Krumhansl, 2004; Krumhansl & Shepard, 1979). The tonic typically sounds at the beginning of a piece of music and, by virtue of its referential role, generally occurs at the end of major phrase boundaries, which provides the listener a sense of appropriate closure (Krumhansl & Kessler, 1982; Schoenberg, 1990). The sense of closure is maximized when the tonic is preceded by the dominant tone (e.g., G in the key of C major) (Schoenberg, 1990). Following the hierarchy of the Western tonal system, the tonic is the most stable tone, whereas the tones outside the scale, also referred to as non-diatonic, are the least stable (Krumhansl & Kessler, 1982; Krumhansl & Shepard, 1979). Both melodies and sequences of chords are arranged according to this hierarchy (Cuddy & Badertscher, 1987; Krumhansl, 2004).

1.3. Studies of musical pitch in typically developing individuals: Development and relationships with cognitive skills

A clear developmental pattern as to pitch-related music skills has been described in TD individuals (see Hannon & Trainor, 2007, and Stalinski & Schellenberg, 2012 for reviews). Pitch discrimination is a first developmental milestone (Jensen & Neff, 1993; Olsho, Schoon, Sakai, Turpin, & Sperduto, 1982). Sensitivity to consonance and dissonance also emerges early in ontogeny (e.g., Trainor et al., 2002). Building on this

skill, knowledge of key membership is acquired (Krumhansl & Keil, 1982; Trainor & Trehub, 1994). Later on, sensitivity to harmonic structure (i.e., "the subtle relationships between notes and chords within a particular key"; Corrigan & Trainor, 2010; p. 200) begins to emerge (Cuddy & Badertscher, 1987; Krumhansl & Keil, 1982; Schellenberg, Bigand, Poulin-Charronnat, Garnier, & Stevens, 2005).

However, as far as we know, there is lack of research on the developmental relationships between cognitive and pitch-related musical skills in TD individuals. Yet, some studies have reported associations between the two types of skills. Pitch processing has been related to matrix reasoning in TD children and adults (Foxton et al., 2003; Norton et al., 2005). Links between pitch-related music skills, auditory short-term memory and working memory have also been established (Anvari, Trainor, Woodside, & Levy, 2002; Huntsinger & Jose, 1991; Wallentin, Nielsen, Friis-Olivarius, Vuus, & Vuus, 2010). In the field of language, both in TD children and adults, pitch processing has been related to phonological awareness and literacy in the native language (Anvari et al., 2002; Barwick, Valentine, West, & Wilding, 1989; Foxton et al., 2003; Lamb & Gregory, 1993), and to phonological skills and linguistic intonation in a foreign language (Delogu, Lampis, & Belardinelli, 2006, 2010; Milovanov, Huotilainen, Välimäki, Esquef, & Tervaniemi, 2008; Milovanov, Pietilä, Tervaniemi, & Esquef, 2010; Slevc & Miyake, 2006). The relationships found between cognitive and pitch-related music skills represent evidence against the views of modularity for music in TD individuals (Schellenberg & Weiss, 2013).

1.4. Studies of musical pitch in Williams syndrome

Following the relevance of pitch in music, a body of research has focused on the assessment of different pitch-related music skills in WS. Levitin (2005) asked individuals with WS (age range not reported) and highly musically trained TD

individuals to discriminate pairs of pitches and found no differences between the groups. Lenhoff et al. (2001a, 2001b) studied absolute pitch, i.e., a rare skill defined as the ability to identify a musical note without a reference pitch (Takeuchi & Hulse, 1993). Five individuals with WS (13-43 years) were assessed on this skill and showed to possess it. Despite its small sample size, this study has been very influential in the characterization of individuals with WS as having an exceptional musical talent (Martínez-Castilla, Sotillo, & Campos, 2013). From a nativist perspective, the fact that individuals with WS can possess absolute pitch in spite of their cognitive impairments has led to support views of innate modularity in music (Lenhoff et al., 2001a, 2001b).

However, other research has shown different results. Martínez-Castilla et al. (2013) found no evidence of absolute pitch in a larger sample of individuals with WS (27 participants aged 12-32 years). When studying pitch discrimination, individuals with WS of different ages (9-15 years in the study of Hopyan, Dennis, Weksberg, & Cytrynbaum, 2001 and 8-41 years in Martens, Reutens, & Wilson, 2010) perform significantly lower than TD individuals matched on chronological age (CA). Likewise, children with WS (8-13 years) discriminate pairs of pitches at the same level as TD peers of the same mental age (MA) (Don, Schellenberg, & Rourke, 1999). From a neuroconstructivist view, these findings indicate that pitch discrimination is not a strength in absolute terms, which, in turn, suggests that this skill is not modular, i.e., it is affected by the cognitive level of individuals with WS (Karmiloff-Smith, Brown, Grice, & Paterson, 2003).

Nevertheless, after using logistic regression, Hopyan et al. (2001) found no relationship between MA and performance on pitch discrimination and this result was considered to support the independence of this skill and cognitive development in WS. However, this result could be accounted for by the way the variable MA was

considered. Hopyan et al. (2001) dichotomized mental ages into older (at least 6.3 years) and younger (less than 6.3 years) for their logistic regression analysis.

Considering both the mean CA of the WS group reported in the study (12 years) and the level of cognitive development typically present in WS (e.g., Martens, Wilson, & Reutens, 2008), most participants should have fallen in the younger group as dichotomized by the authors. Otherwise, the sample would not be representative of the WS population. Either way, the results obtained in the analysis are questionable and no firm conclusions can be drawn from these findings.

Sensitivity to tonality in WS has been analyzed in one study. Martens et al. (2010) presented individuals with WS (8-41 years) with unfinished melodic phrases and asked them to hum the tonic note. Their performance was lower than that of TD individuals matched on CA. Nevertheless, it should be noted that the fact that the participants with WS showed problems for singing the tonic does not necessarily indicate that they were not able to perceive it. Individuals with WS exhibit difficulties in singing in tune (Martínez-Castilla & Sotillo, 2008) and these difficulties may explain the results found by Martens et al. (2010). Perception studies are needed to clarify the extent of sensitivity for tonality in individuals with WS.

Melodic processing has also been investigated in WS. Individuals with WS have been asked to discriminate pairs of melodies that changed in either the global (change in the melody contour) or local (change in the melody interval) levels. Whereas TD individuals show a global precedence (i.e., better performance on the global than local levels), children (8-19 years) (Deruelle, Schön, Rondan, & Mancini, 2005) and adults with WS (16-59 years) (Elsabbagh et al., 2010) exhibit a global deficit that leads them to focus on local cues. According to the neuroconstructivist theory, this finding suggests that atypical processes underlie melodic processing in WS. Interestingly, the same local

processing bias has been identified in different areas of the WS cognitive profile, such as face, spatial, and numerical processing (Annaz, Karmiloff-Smith, Johnson, & Thomas, 2009; Bellugi, Wang, & Jernigan, 1994; Van Herwegen, Ansari, Xu, & Karmiloff-Smith, 2008). This points to common processing strategies across domains (Karmiloff-Smith, 2009).

In stark contrast with the view of musical talent in WS, recent research has studied whether individuals with WS present congenital amusia. In the general population, this disorder is described as a deficit for the processing of musical pitch that cannot be accounted for by brain damage or cognitive impairment (Ayotte, Peretz, & Hyde, 2002). Lense, Shivers, & Dykens (2013) administered to individuals with WS (10-51 years) a diagnostic tool of amusia that assesses the ability to identify pitch anomalies in melodies. Interestingly, a moderate association between performance on the music test and cognitive level was observed. More importantly, Lense et al. (2013) found a higher rate of amusia in WS compared with typical development. Yet, the neural correlates of amusia are similar in both populations (Lense, Dankner, Pryweller, Thornton-Wells, & Dykens, 2014).

In summary, from nativist accounts, it has been claimed that the pitch-related music skills of individuals with WS are preserved and function independently of their cognitive level being furthermore evidence of innate modularity (Lenhoff et al., 2001a, 2001b; Levitin, 2005). However, from neuroconstructivist views, an increasing number of studies suggests that the pitch-related music skills of individuals with WS are not preserved but are affected by their cognitive impairments (Don et al., 1999; Martens et al., 2010). In addition, atypical mechanisms may subserve music processing in WS (Deruelle et al., 2005; Elsabbagh et al., 2010). Furthermore, amusia seems to be particularly prevalent in WS (Lense et al., 2013). In spite of the accumulated evidence,

the idea that individuals with WS have enhanced music skills still persists (e.g., McPherson & Hallam, 2009).

1.5. Scope of the study

Despite the increasing body of evidence that suggests the general cognitive level of individuals with WS has an effect on their pitch-related music skills (e.g., Don et al., 1999; Martens et al., 2010) and, thus, against modularity, further investigation of the relationships between such skills and specific cognitive processes is necessary.

Importantly, it should also be considered that the nativist views that claim innate modularity ignore the crucial role of development in shaping behavioral outcomes (Karmiloff-Smith, 1998a, 2009). Thus, nativist accounts forget that the phenotype in neurodevelopmental disorders does not fully emerge from the outset but develops gradually over time (Thomas et al., 2009). Thereby, as claimed by the neuroconstructivist theoretical view, development should be the key for understanding neurodevelopmental disorders (Karmiloff-Smith, 1998a). Only by assuming a developmental approach will we obtain crucial information regarding the relationships between music and other cognitive processes in WS. However, to our knowledge, no research has previously investigated the development of music skills in this population. Both within and across studies of music cognition in WS, there has been substantial variability in the age range of participants. Moreover, even when the samples have included children, development has not been studied.

Therefore, our research aimed to investigate the developmental trajectories of pitch-related music skills in children with WS to assess both the role of change over time on these skills and the developmental relationships between the music skills and specific cognitive areas. To address this aim, we used the cross-sectional developmental trajectories approach (for simplicity, from now on, developmental trajectories) (Thomas

et al., 2009). In this approach, for both the neurodevelopmental disorder and TD groups, linear regressions are conducted to build separate functions that link task performance on the area of interest with CA or different measurements of MA (as many as are considered relevant to the area under study) (Thomas et al., 2009). Once built, the functions of the groups are contrasted, which enables developmental change and relationships between cognitive areas to be compared across the TD and neurodevelopmental disorder groups (Thomas et al., 2009). This approach has been successfully employed to investigate the development of different areas of the WS cognitive profile, such as face processing and language (e.g., Annaz et al., 2009; Thomas et al., 2010; Van Herwegen, Dimitriou, & Rundblad, 2013).

Under the view that claims pitch-related music skills in WS are preserved, it would be expected that these skills improved with CA in children with WS, which would also be the case in the TD group. However, considering the increasing evidence regarding the effect of cognitive impairments of individuals with WS on their pitch-related music skills, we hypothesized that significant developmental trajectories with CA as a predictor would only be found in the TD group and not in the children with WS. Thus, assuming that pitch-related music skills are related to other cognitive processes, variability in the cognitive level of children with WS would lead to a lack of relationship with CA, i.e., variability in how severely they are affected is not expected to be associated with CA. Instead, in TD children, a significantly close relationship between CA and cognitive development should be found. We also hypothesized that the developmental relationships between the pitch-related music skills and the different cognitive areas included in this study would be atypical in WS, i.e., different than those found in the TD group.

2. Method

2.1. *Participants*

The sample comprised 20 children with WS (10 boys and 10 girls) and 54 TD children (33 boys and 21 girls). The participants with WS presented the clinical phenotype, as well as the genetic markers of the syndrome as demonstrated by Fluorescence In-Situ Hybridization (FISH) tests. No participants had any other clinical diagnoses (as reported by their parents). Participants' parents were administered with a brief questionnaire on the hearing characteristics and musical training of their children. As reported by their parents, no participants in any of the groups suffered from hearing impairment or had received prior formal musical training. The children with WS were recruited through a national Williams Syndrome association and the TD children were recruited through local schools.

All participants were assessed with the following tests of cognitive development: the vocabulary and matrix reasoning subtests of the Wechsler Intelligence Scale for Children-IV (WISC-IV; Wechsler, 2005) or the Wechsler Preschool and Primary Scale of Intelligence-III (WPPSI-III; Wechsler, 2009) (depending on the participant's age) and the forward digit-span and backward digit-span of the WISC-IV (administered to all participants). Measurements of vocabulary and matrix reasoning have often been used to assess cognitive development in WS (e.g., Jarrold, Baddeley, & Hewes, 1998; Mervis, Kistler, John, & Morris, 2012) and, importantly, have also been employed to evaluate claims regarding the separability of music skills and cognition in this population (Don et al., 1999; Hopyan et al., 2001). This explains why such measurements were taken. In addition, as previously mentioned, matrix reasoning has been related to pitch processing in TD individuals (Foxton et al., 2003; Norton et al., 2005), which further justifies the inclusion of this variable. In the same vein, auditory short-term memory and working memory have been linked to pitch-related music skills

in typical development (Anvari et al., 2002; Huntsinger & Jose, 1991; Wallentin et al., 2010). For this reason, participants were assessed with the forward and backward digit-span subtests. From the vocabulary and matrix reasoning subtests, measurements of MA were obtained for each participant. Because the WPPSI-III does not include digit-span subtests and the WISC-IV does not provide measurements of MA for participants younger than 6 years, as done in previous studies (Annaz et al., 2009), raw scores were used as proxies of developmental level in the forward and backward digit-span subtests. Table 1 shows the descriptive characteristics of the WS and TD groups (see Appendix for more details). Note that the sample size varies with the measurement. This is due to floor effects in the WS group (two in vocabulary MA and one in backward digit-span raw score) and floor and ceiling effects in the TD group (one floor effect in the subtest of vocabulary; the remaining cases were ceiling effects). It should also be noted that the age range in the groups was appropriate for the building of developmental trajectories (Thomas et al., 2009).

INSERT TABLE 1 ABOUT HERE

2.2 Materials and procedure

Four tasks were designed for this study and each task evaluated the following pitch-related music skills: pitch discrimination, chord discrimination, dissonance perception, and tonal closure. Figure 1 shows examples of the items in each task.

INSERT FIGURE 1 ABOUT HERE

In the pitch discrimination task, pairs of pitches were presented and the participants were asked whether the two pitches were the same or different. Each pitch lasted 1 second and the pitches ranged from C₄ to C#₅. Half of the pairs were the same and half of the pairs were different. When different, the tonal distance between the two

pitches included ascending and descending intervals from a minor second to a major seventh.

The chord discrimination task comprised pairs of two chords in harmonic relationship (i.e., cadences). The participants had to discriminate whether the pairs were the same or different. The first pair was always a dominant-tonic cadence. In half of the items, the two pairs were the same; in the other half, the two pairs were different. When different, the cadence ended with a scale degree that was not the tonic or with a non-diatonic tone. Each chord lasted 1 second. Within a pair, the two chords were separated by 0.02 seconds and there was a silence of 2 seconds between the two pairs.

In the task of dissonance perception, the participants were presented with tonal sequences of eight chords. In half of the sequences, one chord was replaced with a dissonant cluster, whereas in the other half, the sequences were completely consonant. The participants were asked whether the sequence sounded well or had something that made it sound badly. All tonal sequences remained within a specific key and no modulations (i.e., changes from one key to another) were introduced. The dissonant cluster was formed by changing a chord of the tonal sequence so that its pitches were modified with intervals of major and minor seconds. The cluster was placed between the fourth and seventh chords. Each chord of the sequence lasted 1.45 seconds.

The task of tonal closure was also formed by tonal sequences of eight chords. Following the rules of the Western tonal system (e.g., Schoenberg, 1990), all sequences were composed in a key with one clear tonal center. To avoid confusions regarding the tonal center, no modulations were introduced in the sequences. Within the first seven chords, the key was first established and then a closure cadence was prepared. To do so, the dominant was introduced in the seventh chord so that the sequence could clearly end in the tonic, i.e., the most stable tone and, thus, the tone that provides a sense of

appropriate closure (see section 1.2. *The role of pitch in music*). Finally, after 2 seconds of silence, the final chord was presented. The participants were asked whether the last chord was the appropriate end of the sequence. Half of the sequences ended in the tonic and the other half ended in a tone out of key. Each chord lasted 1.45 seconds.

All materials were composed by an expert musician (the first author) who also played the pitches and chords using a *Samick* piano. The stimuli were digitally recorded (sampling frequency of 22.05 KHz) with a laptop (HP, Intel Pentium M Processor 1.60 GHz 800 MHz, SoundMAX Integrated Digital Audio sound card). The intensity of each pitch or chord was 70 dB, as checked and modified, if necessary, with PRAAT (Boersma & Weenink, 2004).

Each task comprised 20 items: two examples, two practice items (ensuring that the participants understood the task) and 16 experimental items. All participants were individually assessed. The children with WS were tested in a quiet room of their residences and the TD participants were evaluated in a quiet room of their schools. The tasks were presented via loudspeakers at a comfortable listening level. To prevent any effects of boredom or fatigue, the cognitive and pitch-related music tasks were interspersed and breaks were given as needed. The tasks were administered in the following order: pitch discrimination, forward and backward digit-span, tonal closure, vocabulary, dissonance perception, matrix reasoning, and chord discrimination. The parents of the participants provided their written consent for their children to participate in the study after an explanation of its purposes. The study met ethical considerations for the protection of human participants in research and was approved by the corresponding committee.

3. Results

According to the signal detection theory, to account for both the sensitivity to perceiving the pitch-related areas assessed and potential answer biases (Green & Swets, 1966), d-prime scores were calculated for each participant. This measurement takes into consideration the proportions of hits and false alarms (Green & Swets, 1966) and is common in music perception studies (e.g., Elsabbagh et al., 2010; Trainor, 1997; Trainor & Trehub, 1994, 1992). To address occasional extreme values of hits and false alarms (i.e., values of 0 or 1), which arise from the sampling error intrinsic to a limited number of trials and that make it impossible to calculate d-prime scores, the most common solution was followed (Stanislaw & Todorov, 1999). Rates of 0 were replaced with $0.5 / n$ and rates of 1 were replaced with $(n - 0.5) / n$, where n is the number of signal trials (Stanislaw & Todorov, 1999). Mean scores for each group are shown in Table 2.

INSERT TABLE 2 ABOUT HERE

A d-prime score of 0 involves chance performance. To determine whether participant performance on the four tasks was above chance level, separate one-way t tests were conducted for each group in each task. Both groups performed above chance on the pitch discrimination (WS group: $t(19) = 7.86, p < .001, r = .87$; TD group: $t(53) = 28.86, p < .001, r = .97$), chord discrimination (WS group: $t(19) = 5.44, p < .001, r = .78$; TD group: $t(53) = 15.27, p < .001, r = .90$), dissonance perception (WS group: $t(19) = 5.81, p < .001, r = .80$; TD group: $t(53) = 16.01, p < .001, r = .91$) and tonal closure tasks (WS group: $t(19) = 4.02, p = .001, r = .68$; TD group: $t(53) = 9.08, p < .001, r = .78$).

Separate one-way t tests were also conducted to test whether there were floor or ceiling effects on the pitch-related music tasks. A d-prime score of -3.07 corresponds to a floor effect (resulting from no hits and a maximum number of false alarms) whilst a d-

prime of 3.07 involves a ceiling effect (maximum number of hits and no false alarms). There were no floor effects on any task in any of the groups (pitch discrimination: WS group: $t(19) = 20.02, p < .001, r = .98$; TD group: $t(53) = 62.88, p < .001, r = .99$, chord discrimination: WS group: $t(19) = 19.77, p < .001, r = .98$; TD group: $t(53) = 38.95, p < .001, r = .98$, dissonance perception: WS group: $t(19) = 18.44, p < .001, r = .97$; TD group: $t(53) = 42.40, p < .001, r = .99$, tonal closure: WS group: $t(19) = 20.37, p < .001, r = .98$; TD group: $t(53) = 29.07, p < .001, r = .97$). No ceiling effects were found either (pitch discrimination: WS group: $t(19) = -4.03, p < .001, r = .68$; TD group: $t(53) = -5.16, p < .001, r = .58$, chord discrimination: WS group: $t(19) = -8.88, p < .001, r = .90$; TD group: $t(53) = -8.41, p < .001, r = .76$, dissonance perception: WS group: $t(19) = -6.82, p < .001, r = .84$; TD group: $t(53) = -10.38, p < .001, r = .82$, tonal closure: WS group: $t(19) = -12.33, p < .001, r = .94$; TD group: $t(53) = -10.91, p < .001, r = .83$).

As previously mentioned (see section 1.4. *Studies of musical pitch in Williams syndrome*), the prevalence rate of amusia in individuals with WS is higher than that in the TD population (Lense et al., 2013). As shown in Figure 2, in this study no participant with WS performed at floor. Thus, none of them seemed to have amusia.

INSERT FIGURE 2 ABOUT HERE

3.1. Developmental trajectories for the typically developing group

Developmental trajectories for the TD group were built by running separate linear regression analyses for the performance on each task where CA, vocabulary MA, matrix reasoning MA, forward digit-span score, or backward digit-span score were entered as predictors. Results are presented in Table 3.

INSERT TABLE 3 ABOUT HERE

As shown in Table 3, in the TD group, the performance on all pitch-related music tasks significantly improved with CA, vocabulary MA, matrix reasoning MA,

forward digit-span, and backward digit-span scores. These developmental trajectories are shown in Figure 3. For the sake of clarity, individual data points are not shown.

INSERT FIGURE 3 ABOUT HERE

3.2. *Developmental trajectories for the Williams syndrome group*

Developmental trajectories were also built for the children with WS. Results are shown in Table 4.

INSERT TABLE 4 ABOUT HERE

As can be seen in Table 4, in contrast with the TD group, few studied variables were related to the tasks in the WS group. CA only predicted performance on the chord discrimination task. Matrix reasoning MA was a significant predictor only for the chord discrimination and tonal closure tasks. Finally, the backward digit-span score significantly predicted performance on pitch discrimination, chord discrimination, and tonal closure. It should be mentioned that nonlinear functions did not fit the data (i.e., logarithmic, exponential, growth). All developmental trajectories of the WS group are shown in Figure 4, where, for the sake of clarity, individual data points are not presented.

INSERT FIGURE 4 ABOUT HERE

The lack of significant relationships between the remaining tasks and predictors can be because of the existence of a real non-systematic relationship (i.e., random performance with respect to the predictors) or a zero trajectory (i.e., the scores on the task are horizontally distributed so that performance does not change with age) (Thomas et al., 2009). These two possibilities can be distinguished if the scores are transformed by a 45° anti-clockwise rotation (see Thomas et al., 2009 for full details). Once the rotation has been performed, the R^2 value of the randomly distributed data remains non-significant and close to zero, whereas the R^2 value of a zero trajectory increases and

becomes highly significant (Thomas et al., 2009). This method was therefore applied to our data. Non-systematic relationships were found with CA as the predictor for the pitch discrimination, dissonance perception, and tonal closure tasks ($F(1, 18) = 3.61, p = .07, R^2 = .17$; $F(1, 18) = 0.41, p = .53, R^2 = .02$; and $F(1, 18) = 0.12, p = .07, R^2 = .00$, respectively). Instead, zero trajectories were found with vocabulary MA as the predictor for all tasks (pitch discrimination: $F(1, 17) = 59.69, p < .001, R^2 = .78$; chord discrimination: $F(1, 17) = 7.06, p = .02, R^2 = .29$; dissonance perception: $F(1, 17) = 13.10, p = .002, R^2 = .44$; and tonal closure: $F(1, 17) = 7.27, p = .02, R^2 = .30$). Zero trajectories were also found with matrix reasoning MA for the two tasks where no relationships had been previously found (pitch discrimination: $F(1, 18) = 96.86, p < .001, R^2 = .84$; and dissonance perception: $F(1, 18) = 47.64, p < .001, R^2 = .73$). The same result was obtained with the forward digit-span score for all tasks (pitch discrimination: $F(1, 18) = 36.41, p < .001, R^2 = .67$; chord discrimination: $F(1, 18) = 8.68, p = .009, R^2 = .33$; dissonance perception: $F(1, 18) = 16.29, p = .001, R^2 = .48$; and tonal closure: $F(1, 18) = 7.32, p = .01, R^2 = .29$). Finally, a zero trajectory was also observed with backward digit-span as the predictor of performance on the dissonance perception task ($F(1, 16) = 31.50, p < .001, R^2 = .66$).

In summary, once the rotation method was used (Thomas et al., 2009), with the exception of CA where non-systematic relationships were found, zero trajectories were obtained with vocabulary MA, matrix reasoning MA, forward digit-span score, and backward digit-span score on the tasks in which the initial regression analyses had yielded non-significant results.

3.3. Comparison of developmental trajectories between the Williams syndrome and typically developing groups

Between-group comparisons of developmental trajectories enable the assessment of whether the intercepts and gradients of the linear regressions obtained for each group are the same (Thomas et al., 2009). The intercept refers to the onset of the developmental trajectory, whereas the gradient represents the rate of development. Developmental delays and slower or faster trajectories can then be inferred when between-group differences are found in the intercept or gradient, respectively (Thomas et al., 2009). These comparisons can only be conducted when significant linear trajectories (i.e., significant linear regressions) have previously been found for the groups under comparison (Thomas et al., 2009). Therefore, we could only compare the developmental trajectories (i.e., their intercepts and gradients) of the WS and TD groups for the following tasks and predictors: the pitch discrimination task and backward digit-span score, the chord discrimination task and CA, matrix reasoning MA, or backward digit-span score, and the tonal closure task and matrix reasoning MA, or backward digit-span score.

In all cases, to compare the linear regressions of the WS and TD groups, we used ANCOVA tests (see Thomas et al., 2009 for details). To compare the intercept values at the first point of measurement (i.e., the earliest age measured), we re-scaled all predictors to the youngest age or score measured in the WS or TD groups. In this way, the potential difference in the onset of development is shown by the main effect of the variable Group (WS vs. TD groups). The main effect of the covariate (CA, vocabulary MA, matrix reasoning MA, forward digit-span score, or backward digit-span score) tests whether, with the groups combined, performance is significantly predicted by the covariate. However, we were interested in assessing whether the gradient of the linear regressions of the WS and TD groups was different. Therefore, we also included the interaction term of Group x Predictor in the model.

For the pitch discrimination task, no significant effect was found for the variable Group ($F(1, 68) = 1.75, p = .19, \eta_p^2 = .025$). The covariate backward digit-span score yielded significant results ($F(1, 68) = 12.92, p = .001, \eta_p^2 = .16$). However, the interaction term was non-significant ($F(1, 68) = 3.68, p = .06, \eta_p^2 = .05$).

For the chord discrimination task, when CA was considered the covariate, a significant main effect of Group was found; thus, the performance was lower for the WS group compared with the TD group ($F(1, 70) = 7.93, p = .006, \eta_p^2 = .10$). Therefore, a developmental delay was identified in the WS group. The covariate was significant ($F(1, 70) = 15.59, p < .001, \eta_p^2 = .18$). However, the interaction between Group and CA was non-significant ($F(1, 70) = 0.09, p = .76, \eta_p^2 = .001$). The analysis was also independently conducted with matrix reasoning MA and the backward digit-span score as covariates. With matrix reasoning MA, no significant effects were found for Group or the interaction between Group and the covariate ($F(1, 61) = 1.78, p = .19, \eta_p^2 = .03$; and $F(1, 61) = 3.41, p = .07, \eta_p^2 = .05$, respectively). Nevertheless, the covariate itself was significant ($F(1, 61) = 8.01, p = .006, \eta_p^2 = .12$). The same results were found when the backward digit-span score was considered the covariate (Group: $F(1, 68) = 2.68, p = .11, \eta_p^2 = .04$; covariate: $F(1, 68) = 10.14, p = .002, \eta_p^2 = .13$; and interaction term: $F(1, 68) = 2.61, p = .11, \eta_p^2 = .04$).

Finally, for the tonal closure task and the matrix reasoning MA, Group and the interaction term were not significant ($F(1, 61) = 0.001, p = .97, \eta_p^2 = .00$; and $F(1, 61) = 3.64, p = .06, \eta_p^2 = .06$, respectively). However, the covariate matrix reasoning MA was significant ($F(1, 61) = 10.38, p = .002, \eta_p^2 = .15$). The same results were found for the tonal closure task and the backward digit-span score as the covariate (Group: $F(1, 68) = 0.002, p = .96, \eta_p^2 = .00$; covariate: $F(1, 68) = 8.53, p = .005, \eta_p^2 = .11$; and interaction term: $F(1, 68) = 0.52, p = .48, \eta_p^2 = .008$).

In summary, there was a significant effect of Group only for the chord discrimination task with CA as the covariate. For all the tasks, there was a significant effect of the covariates considered. No significant results were found for any of the interaction terms.

3.4. Comparison of developmental trajectories of the pitch-related tasks in each group

A repeated measures design was used to compare the developmental trajectories of the different pitch-related tasks in each group. In this design, the covariate (i.e., CA, vocabulary MA, matrix reasoning MA, forward digit-span score, or backward digit-span score) and the within-subjects factor (i.e., the pitch-related tasks) are orthogonal; therefore, the main effect of the within-subjects factor is independent of the potential effect of the covariate (Thomas et al., 2009). As a consequence, the analysis must be conducted in two phases: first, the covariate is omitted and the within-subject effect is studied by means of a repeated measures ANOVA; second, the covariate is added and an ANCOVA is conducted (see Thomas et al., 2009 for a detailed explanation in this regard).

As previously seen, in our study, only a few covariates predicted performance on the pitch-related tasks and, in one particular task, dissonance perception, no significant relationships were found with any of the covariates. Therefore, to compare the developmental trajectories of the different pitch-related tasks in each group, we only conducted the first phase of the analysis, i.e., the repeated measures ANOVA with Task as the within-subject factor.

In the TD group, the main effect of Task was significant ($F(1, 53) = 85.13, p < .001, \eta_p^2 = .62$). Bonferroni pairwise comparisons showed that performance on pitch discrimination was higher than that on chord discrimination ($CI_{.95} = 0.37$ (lower) 0.88 (upper), $p < .001$), dissonance perception ($CI_{.95} = 0.48$ (lower) 1.00 (upper), $p < .001$),

and tonal closure ($CI_{.95} = 0.83$ (lower) 1.59 (upper), $p < .001$). In turn, the results on chord discrimination were higher than those on tonal closure ($CI_{.95} = 0.24$ (lower) 0.93 (upper), $p < .001$), but at the same level as the ones found for dissonance perception ($CI_{.95} = -0.19$ (lower) 0.42 (upper), $p = 1.00$). Finally, the TD individuals performed better on the dissonance perception task than on the tonal closure task ($CI_{.95} = 0.05$ (lower) 0.89 (upper), $p = .02$).

In the WS group, the pattern of results was different. Performance on pitch discrimination was higher than that on chord discrimination ($CI_{.95} = 0.19$ (lower) 1.44 (upper), $p = .006$) and tonal closure ($CI_{.95} = 0.48$ (lower) 1.98 (upper), $p = .001$). However, the results were not significantly different between pitch discrimination and dissonance perception ($CI_{.95} = -0.34$ (lower) 1.48 (upper), $p = .47$). Moreover, no significant differences were found between any other pair of tasks. Thus, the individuals with WS performed similarly on chord discrimination and dissonance perception ($CI_{.95} = -0.95$ (lower) 0.46 (upper), $p = 1.00$), chord discrimination and tonal closure ($CI_{.95} = -0.12$ (lower) 0.95 (upper), $p = .21$), and dissonance perception and tonal closure ($CI_{.95} = -0.13$ (lower) 1.45 (upper), $p = .15$).

In summary, in the TD group, there was a clear developmental pattern. Pitch discrimination was the easiest task, chord discrimination and dissonance perception followed, and tonal closure was the most difficult task. In the WS group, although pitch discrimination was easier than chord discrimination and tonal closure, no other differences were found between the tasks.

4. Discussion

In this study, we evaluated the developmental trajectories of different pitch-related music skills in children with WS compared with TD children. First, we investigated the relationships between the pitch-related music skills and CA. As

hypothesized, the results were significant only for the TD children. Thus, while the pitch-related music skills of the TD children improved with CA, no-systematic relationships were found between CA and pitch discrimination, dissonance perception, or tonal closure, respectively, in the children with WS. This lack of relationship is a marker of atypical development (Thomas et al., 2009). It also constitutes further evidence against the view that pitch-related music skills in WS are preserved and represent an area of strength in absolute terms (Lenhoff et al., 2001a, 2001b; Levitin, 2005). Only for the chord discrimination task was there a significant relationship with CA in WS. However, when the developmental trajectories of the WS and TD groups were compared in this task, a developmental delay was identified in the WS group. This finding provides additional evidence against the idea that pitch-related music skills represent an absolute strength in WS. Nevertheless, we should note that, in light of this study, it is not clear why the skill for chord pair discrimination improved with CA in children with WS. Indeed, considering the lack of systematic relationships between CA and the remaining pitch-related music skills, the significant relationship found with chord discrimination may be surprising. Further research should clarify the reasons why the relationships between CA and pitch-related music skills differed between the tasks included in the study.

We also investigated the developmental relationships between different cognitive areas and the pitch-related music skills. In the TD children, significant developmental trajectories were found with all cognitive areas included in the study, i.e., vocabulary, matrix reasoning, forward digit-span, and backward digit-span. These results are consistent with those found in prior studies (Anvari et al., 2002; Foxton et al., 2003; Huntsinger & Jose, 1991; Norton et al., 2005; Wallentin et al., 2010). However, in the children with WS, few cognitive variables were related to the pitch-related music

skills. Backward digit-span predicted performance on pitch discrimination, chord discrimination and tonal closure, and matrix reasoning predicted the scores obtained on the chord discrimination and tonal closure tasks. In contrast, vocabulary and forward-digit span were not significant predictors in any music task.

The significant relationships found with backward digit-span can be explained by considering that this subtest measures auditory working memory and the pitch-related music skills in which the relationships were found require this cognitive process. Thus, the pitch discrimination, chord discrimination and tonal closure skills involve retaining and comparing different auditory patterns in memory (pitches, chords, and sequences of chords, respectively), i.e., auditory working memory. A similar argument can be used to explain why matrix reasoning predicted performance on chord discrimination and tonal closure. The matrix reasoning subtests used in this study (those of WISC-IV and WPPSI-III) require participants to establish a logic relationship between different items and, on the basis of the relationship established, to answer a question (e.g., which is the following pattern in the sequence, which item completes the pairs of pictures presented). In chord discrimination, performance is facilitated when cadential relationships between chord pairs are established. In turn, tonal closure requires the comprehension of the harmonic relationships of chord sequences. Therefore, the cognitive skills involved in matrix reasoning are also involved in the chord discrimination and tonal closure skills, which explains the significant developmental trajectories found. In contrast to chord discrimination and tonal closure, pitch discrimination does not involve the establishment of a logic or meaningful relationship between the two pitches presented. This argument offers a likely explanation as to why no significant developmental relationships were found between pitch discrimination and matrix reasoning.

As aforementioned, in WS, vocabulary and forward-digit span did not predict performance on any pitch-related music tasks. Following the same reasoning as previously discussed, it should be considered that vocabulary is not involved in the pitch-related music skills of the study. This could account for the lack of relationships found. Similarly, although forward-digit span measures auditory short-term memory, as previously explained, the pitch-related music skills of the study involve active working memory and not mere storing. This factor may explain why forward-digit span was not a significant predictor.

While some developmental trajectories were found in WS for pitch discrimination, chord discrimination and tonal closure, none of the cognitive variables in this study predicted performance on the dissonance perception task. In this task, the participants with WS tended to respond as soon as they perceived the dissonant cluster instead of answering at the end of the chord sequence. For this reason, it may be considered that the WS group performed on dissonance perception as if it were an online task. Online tasks require less cognitive processes to be solved (Karmiloff-Smith et al., 1998). In this case, solving the task would only require the participants to identify a dissonance, without the use of their auditory working memory (i.e., without storing and comparing auditory patterns) or the establishment of meaningful relationships between the chords. This factor would explain why no significant developmental trajectories were found between this task and backward-digit span or matrix reasoning in the WS group. In contrast with dissonance perception, pitch discrimination, chord discrimination, and tonal closure were necessarily off-line tasks that could not be solved via online processing.

As shown, significant developmental trajectories were found in WS only for the pitch-related music skills in which some of the specific cognitive processes assessed in

this study were involved. Interestingly, in such cases, when the trajectories of the WS and TD groups were compared, the developmental relationships between the cognitive and pitch-related music skills were not significantly different. Therefore, the performance on the pitch-related music skills was in line with the development of the cognitive skills deemed relevant for these music skills. This finding also represents evidence against the claims that in WS music constitutes a module independent of cognition (e.g., Levitin & Bellugi, 1998; Lenhoff et al., 2001a, 2001b).

The lack of significant differences between the previously described developmental trajectories in the WS and TD groups might lead to think that the development of pitch-related music skills is typical in WS. However, prior to drawing such conclusion, we should consider an important difference in the results of the groups. As previously discussed, in contrast with the TD children who exhibited significant developmental relationships between all pitch-related music skills and cognitive areas included in the study, in the children with WS, only two cognitive variables (i.e., backward-digit span and matrix reasoning) were related to some of the pitch-related music skills. This is therefore a marker of atypicality. In TD children, the different cognitive skills develop hand in hand, increasing, in turn, with CA. This would explain why significant developmental trajectories were found for the TD group with all the predictors included in the study. In WS, instead, the cognitive profile is uneven and the different cognitive areas do not always develop in synchrony (Bellugi et al., 2000; Jarrold et al., 1998), which would account for the results of the WS group. Nevertheless, it should be noted that this unevenness can represent a means to ascertain subtle associations between different skills which would be otherwise difficult to investigate in TD children (Van Herwegen, Riby, & Farran, 2015). Indeed, in our study, the results found in the WS group have informed us regarding cognitive skills specifically involved

in different pitch-related music skills (e.g., auditory working memory in pitch discrimination, chord discrimination, and tonal closure; matrix reasoning in chord discrimination and tonal closure). It might be argued that the results of the WS group may instead be an artifact of the smaller sample size of this group as compared to the TD group. However, this would not explain why only some cognitive variables were found as significant predictors in the children with WS.

Consistent with their uneven cognitive profile, the zero trajectories found in the group with WS represent another marker of atypical development. While the scores on the pitch-related music tasks always improved with cognitive development in the TD children, in the participants with WS, in many cases, cognitive development (e.g., vocabulary development or auditory short-term memory development) was not paired with an improvement in the pitch-related music skills. This finding would involve stabilization in the developmental level of the pitch-related music skills of children with WS when these skills are compared with their progressing developmental level in other cognitive areas.

When the developmental trajectories of the different pitch-related music skills of the study were compared, the results also differed between the WS and TD groups. In the TD children, a clear developmental pattern was identified in which pitch discrimination developed first, chord discrimination and dissonance perception followed, and tonal closure developed last. This pattern is consistent with the literature regarding the development of pitch-related music skills (see section 1.3. *Studies of musical pitch in typically developing individuals: Development and relationships with cognitive skills*). Thus, pitch discrimination occurs very early in ontogeny and continues to develop during childhood (Jensen & Neff, 1993). Chords are formed by individual pitches; thus, chord discrimination should follow pitch perception. On the basis of the

relative ratios between pitches, consonance (simple ratios) and dissonance (complex ratios) arise and the perception of these phenomena is also an early developmental milestone (e.g., Trainor et al., 2002). Sensitivity to consonance and dissonance subsequently functions as a crucial building block for learning the pitch structure of the Western musical system, i.e., the hierarchical pitch organization within scales or tonalities (Hannon & Trainor, 2007).

The developmental pattern in the children with WS was very different. Pitch discrimination developed earlier than chord discrimination and tonal closure. However, no significant differences were found between chord discrimination, dissonance perception, and tonal closure. Therefore, in the WS group, although performance was above chance level in all music tasks, chord discrimination and dissonance perception did not precede tonal closure. This finding is surprising considering tonal closure would involve the prior acquisition of chord discrimination and dissonance perception. Thus, of these skills, tonal closure develops the latest in TD children (Hannon & Trainor, 2007). This result suggests that the underlying processes employed by children with WS to perceive the tonal hierarchies of the Western musical system (and thus to solve the tonal closure task) could be different from the processes employed by TD children. Future research should further investigate this issue.

As previously mentioned, in this study we have found new evidence against the views of innate modularity for music in WS. Similar conclusions have been reached in prior studies in TD individuals (e.g., Patel, 2008; Schellenberg & Weiss, 2013; but see Peretz & Coltheart, 2003, for a proposal of modularity) and our results in the TD group are consistent with them. However, it is important to note that the results found in WS should not be taken to support theories of cognitive organization in typical development. The atypicalities found in the development of pitch-related music skills in

WS show that this neurodevelopmental disorder should not be considered a window on the cognitive architecture of TD individuals (Karmiloff-Smith, 1998b).

An important finding of this research is the set of atypicalities observed in the development of pitch-related music skills in children with WS. Although, to our knowledge, no prior research has reported atypical development of music skills in WS, previous studies have found other atypicalities in the musical phenotype of the syndrome. Even though individuals with WS do not have enhanced music skills (Lense et al., 2013; Martínez-Castilla & Sotillo, 2008), they show greater musicality. That is, in comparison with TD individuals or those with other neurodevelopmental disorders, individuals with WS present heightened interest and emotional responsivity to music, as reported by parents and evidenced by electroencephalogram and electrodermal activity (Hopyan et al., 2001; Järvinen et al., 2015; Lense, Gordon, Key, & Dykens, 2014; Levitin et al., 2004). In addition, atypical links have been found between musicality and socio-emotional sensitivity (Ng, Lai, Levitin, & Bellugi, 2013). Different patterns of neural activation in the presence of music have also been found in WS. A wider network, including the amygdala, is activated, which could account for the high proclivity to music of individuals with WS (Levitin et al., 2003; Thornton-Wells et al., 2010). As far as we know, no prior research has studied how these atypical neural underpinnings relate to the development of music skills in general or pitch-related music skills in particular in WS. Future studies should focus on this topic.

Before concluding, some methodological issues should be considered. As previously mentioned, chance performance and floor or ceiling effects were ruled out and therefore cannot explain the results of the study. The sample size of the groups was also appropriate for building developmental trajectories (see, e.g., Annaz et al., 2009; Naylor & Van Herwegen, 2012; Thomas et al., 2010; Van Herwegen et al., 2013).

However, it should be noted that the number of participants in the WS group was smaller than that in the TD group. Although this is common in the literature on WS due to the low incidence of the syndrome and the subsequent difficulties for recruiting participants, it also involves lower statistical power for the analyses of this group, which may represent a limitation of the study. It should also be considered that, even though the use of developmental trajectories represents a sound method for studying the development of pitch-related music skills (or any other skills) in WS, future longitudinal studies should validate the results of this research.

5. Conclusions

To conclude, our investigation of the developmental trajectories of pitch-related music skills in children with WS has contributed to the theoretical debates regarding music cognition in WS. We have identified developmental relationships between several pitch-related music skills and related cognitive processes. We have also reported clear markers of atypicality in the development of pitch-related music skills in children with WS. Thus, we have found a general lack of relationship between these skills and CA, uneven associations between cognitive and pitch-related music skills, zero trajectories, and an atypical pattern of pitch-related music skill development. These findings suggest that the developmental pathways of pitch-related music skills in WS are different from those of TD children. Overall, our results stand in contrast with the nativist views of modularity for music in WS (e.g., Levitin & Bellugi, 1998; Lenhoff et al., 2001a; Wengenroth et al., 2010). Instead, they are consistent with the neuroconstructivist accounts (e.g., Elssabagh et al., 2010; Karmiloff-Smith, 2009). Overall, the findings highlight the relevance of considering development in the investigation of developmental disorders.

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